ISSN - 2250-1991 | IF : 5.215 | IC Value : 79.96

Original Research Paper Subject: Medicine

UNILATERAL LUNG AGENESIS- AN ADULTHOOD PRESENTATION

Dr.Pankaj Hans	Assistant Professor, Department of General Medicine, Patna Medical College, Patna 800004
Dr.Rahul Kumar	Assistant Professor, Department of Geriatric Medicine, Patna Medical College, Patna 800004
Dr.Sushant Kumar Pathak	Post Graduate student, Department of General Medicine, Patna Medical College, Patna 800004
Dr.Kumar Himanshu	Post Graduate student, Department of General Medicine, Patna Medical College, Patna 800004
Dr.Manjunathswamy	Post Graduate student, Department of General Medicine, Patna Medical College, Patna 800004
Dr.Gaurav Kumar	Post Graduate student, Department of General Medicine, Patna Medical College, Patna 800004

ABSTRACT

Agenesis of lung being a rare entity was first described by DePozze in 1673 and later in India by Muhamed, both during an autopsy.It has the prevalence of 34 per million live birth .Bilateral agenesis is fatal and the unilateral presentation is more common with the absence of left side of lung.We are reporting here a case of left Lung agenesis in a 21 yr old girl who presented with complains of dry cough and mild breathlessness in our OPD ,and later proceeding with the Chest X Ray ,HRCT thorax and fibreoptic bronchoscopy we reached the diagnosis.

KEYWORDS

INTRODUCTION:-

Lung agenesis(1) is although a less common developmental anomaly of lung tissue in comparison to others like bronchogenic cysts, pulmonary sequestration etc(4). Except in cases with bilateral agenesis most patients present with recurrent chest infections since early childhood. Diagnosis in adults is very rare(2). The oldest patient cited by OYAMADA et al was 72 yrs old(6). We hereby present a case of pulmonary agenesis diagnosed in adulthood**.**

CASE REPORT

A 21 yr old female patient from rural Bihar, a farmer by occupation presented in our OPD with dry cough and progressive shortness of breath for 18-20days.She had no any history of allergy, paroxysmal attacks of breathlessness, Tuberculosis, familial pulmonary disorders, weight loss, Hemoptysis or prior hospital admissions.

On general examination, she was an average built alert ,conscious and cooperative patient having no pallor, icterus, cyanosis, clubbing, edema and lymphadenopathy. BP was 110/84 in sitting position in bilateral upper limb and Pulse rate was 68/minute regular in rhythm. Her remarkable general examination finding was only tachypnea.

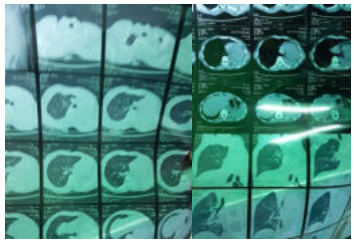
SYTEMIC EXAMINATION:

Her upper Respiratory tract was apparently normal. On examining lower respiratory tract, She had Pectus excavatum, diminished movement of left whole hemi thorax, trachea shifted to left with a respiratory rate of 36/minute with no intercostals suction, no obvious vertebral deformity. Dull percussion was noted in whole left hemi thorax accompanying prolonged expiration with decreased intensity of vesicular breath sound in all over of right hemithorax.We noted occasional rhonchi in right hemithorax.
Other system examination was apparently normal.

INVESTIGATIONS:

Routine blood reports:
Hb-10.8
WBC-6,500 (Neutrophils-71%, lymphocytes-22%, eosinophils-6%)
Serum Ig E slightly raised, serum Ig G and LDH normal
Her induced sputum examination was negative for AFB, CBNAAT and Gram stain. No growth was found in culture too.

Chest X-ray and HRCT-She had opaque homogenous opacity in left hemi thorax with ipsilateral mediastinal shift. HRCT shows frank absence of left whole lung with its supplying vasculature and bronchus. It showed herniating Right lung into left side as well.



Pulmonary function test was of restrictive pattern predominantly. Fibreoptic bronchoscopy was done showing no left bronchus although right side was totally normal.
USG whole abdomen and 2D echo with Doppler was found to be normal.

So we finally diagnosed it a case of Left pulmonary agenesis with URTI which responded with antibiotics, antihistaminic and bronchodilators. She improved gradually and is in our follow up since then.

DISCUSSION:

Unilateral lung agenesis may have either symptomatic or asymptomatic presentation(3).50-80%cases are associated with cardiovascular, gastrointestinal, genitourinary, vertebral anomaly. It presents as a developmental anomaly occurring in 4th-5th week of embryonic phase of pulmonary development before pseudo glandular period when primitive lung are starting to form as a diverticulum protruding from foregut(5). Although the exact etiology is not known, various causative factors have been postulated like deficiency of Vit A, Chromosomal aberrations, intrauterine infections etc. More commoner agenesis is with Left hemi thorax but since these have less associated other anomalies, prognosis is better compared to right.

Pulmonary agenesis has been classified into three categories depending upon the stage of development of primitive lung bud. It was first proposed by SCHNEIDER AND SCHWATBE then later modified by BOYDEN.

1.**PULMONARY AGENESIS**-Whole pulmonary parenchyma, bronchus as well as pulmonary artery is absent.

2. **PULMONARY APLASIA**-Rudimentary bronchus with complete absence of pulmonary parenchyma

3.**PULMONARY HYPOPLASIA**-Variable amount of pulmonary parenchyma, bronchi and pulmonary vessels are present.
Important associations are as illustrated below(7):

A. Cardiovascular –PDA, Patent foramen Ovale

B. GIT- Tracheo -esophageal Fistula, Duodenal Artesia

C. Renal –Horshoe Kidney

D. Vertebral –VACTERL (Vertebral anomaly, Anal atresia, cardiac structural defect, Tracheo-oesophgeal fistula, esophageal atresia and limb aplasia)

A rare association with MRKH syndrome has also been reported.

CONCLUSION

We conclude that agenesis of lung can manifest as simple as URTI in adulthood, it may clinically mimic as pleural effusion or unilateral lung collapse but a little high degree of suspicion is needed to go further for its diagnosis.

References :

1. Dutta D,Karlekar A,SaxenaR:Agenesis of lung,Ann Card Anaesthesia, 2013;16(2),151-52
2. Roy PP,Datta S,Sarkar et al;Unilateral pulmonary agenesis presentic in adulthood, Respiratory medicine case report 2012,5;81-83
3. Savas S,Hacieuliyagil,Gunen H et al:Asymptomatic pulmonary agenesis,over experience with two cases,Turkish Respiratory Journal2006;7(1),31-33
4. Koseoglu N,Uncan ES,Cavdar C. Respiratory medicine Extra 2005;vol 1.Issue 4110-112
5. Viora E,Sciarrone A,Bastonero S,Errante G,Campogrande M.Prenatal diagnosis of unilateral pulmonary agenesis in 2nd trimester.Ultrasound Obstet Gyanecol2002;19;206-7
6. Oyamada A,Gasul BM,Holinger PM,Agenesis of lung,Am J Dis child 1953;85:182-7
7. Boyden E Developmental Anomalies AM J Surg 1955;89 ;79-88